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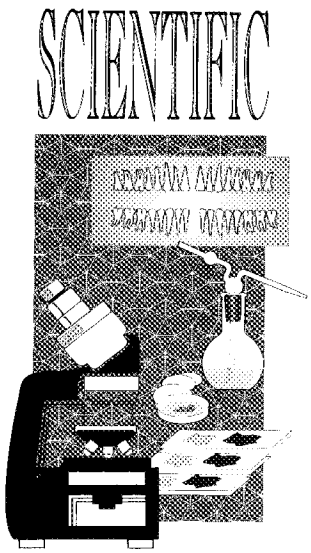

Osteosarcoma and Fibrous Dysplasia: Radiographic Features In the Differential Diagnosis: A Case Report

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ABSTRACT

A case of osteosarcoma misdiagnosed as fibrous dysplasia is presented to demonstrate the importance of an integrated diagnostic approach to oral lesions. The clinical and radiographic differences between fibrous dysplasia and osteosarcoma are reviewed.

SOMMAIRE

Dans cet article, on présente un cas de sarcome ostéogénique (ostéosarcome), diagnostiqué à tort comme une dysplasie fibreuse, afin de démontrer l'importance d'une approche intégrée pour diagnostiquer les lésions buccales de ce genre. De plus, on y examine les différences cliniques et radiologiques entre l'ostéosarcome et la dysplasie fibreuse.

Introduction

Monostotic fibrous dysplasia is a benign, slowly growing lesion that most often occurs in children. It usually remains active until the end of skeletal growth, although Henry[1] reports recrudescence during pregnancy. There is no sex predilection.[2] It can occur in either jaw and is most often found in the molar/premolar region of the maxilla.[3] In the jaws, it is usually monostotic and unilateral.[3] Enlargement of the jaw typically appears as a relatively smooth, fusiform expansion, with the overlying mucosa being intact and normal in appearance (**Fig. 1**). Usually, the only symptom is swelling or facial asymmetry. Teeth may occasionally be displaced or prevented from erupting.

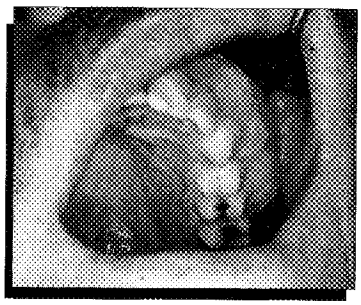

Fig. 1: *Clinical appearance of maxillary fibrous dysplasia demonstrating the marked, fusiform expansion of the buccal cortex, and intact normal appearing mucosa.*

Microscopically, the lesion consists of a cellular fibrous stroma containing irregular, sometimes interconnecting, islands of bone that usually lack osteoblastic rimming. The cells do not exhibit malignant features and mitotic activity is not prominent. The lesional tissue fuses directly with surrounding bone without the formation of a fibrous capsule. Unless it is necessary for cosmetic or functional reasons, no treatment is required.

Osteosarcoma is a malignant, often fatal disease of bone that occurs more often in young adult males.[4,5] In the jaws, osteosarcoma tends to occur somewhat later than in the long bones, usually in the third[6] or fourth[5] decade. The initial symptom is often asymptomatic, localized swelling that exhibits a variable growth rate. As the lesion progresses, pain, loosening of teeth, paresthesia, toothache and gingival bleeding may also occur. Microscopically, there are several variants of the malignancy. All exhibit areas of bone or osteoid associated with malignant osteoblasts or less differentiated cells. The tumors may show areas of malignant fibrous or cartilaginous tissue. Mitotic activity is usually present. The neoplasm characteristically invades into surrounding structures, often perforating cortical plates. Treatment consists of resection accompanied by chemotherapy or radiotherapy. The five-year survival rate for osteosarcoma of the jaws is reported to be 32 per cent.[4] For tumors that are specific to the maxilla, however, the survival rate is reported to be 25.8 per cent.[7]

Both monostotic fibrous dysplasia and osteosarcoma may affect the jaw bones, producing bony swellings. Initially, both lesions may be asymptomatic, but their eventual biologic behaviors are

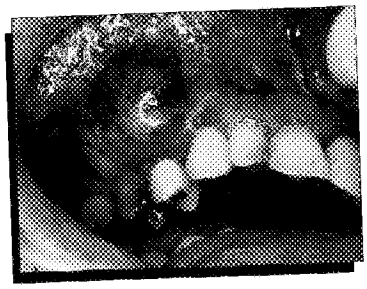

Fig. 2: Clinical appearance of osteosarcoma demonstrating the non-ulcerated mucosa and lobulated appearance.

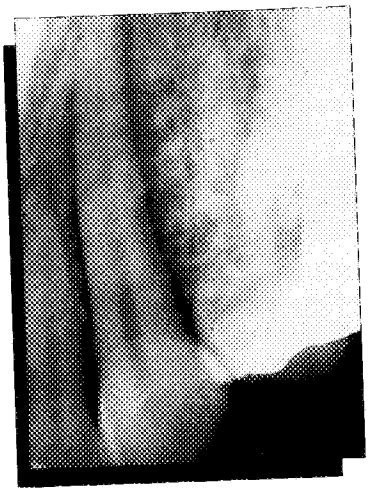

Fig. 3: Maxillary periapical view of osteosarcoma demonstrating the "moth-eaten" poorly-delineated bone pattern and irregular widening of the periodontal ligament space about the cuspid.

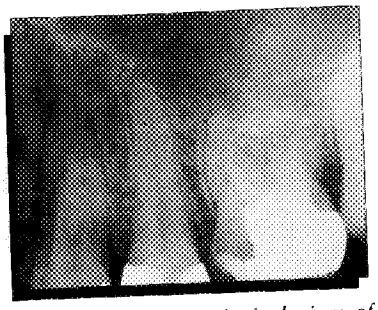

Fig. 4: Maxillary periapical view of osteosarcoma demonstrating the appearance of calcification within the altered bone pattern.

Fig. 5: Overexposed copy film of lateral maxillary occlusal view demonstrating irregular osseous spiculation extending at right angles from the buccal cortex.

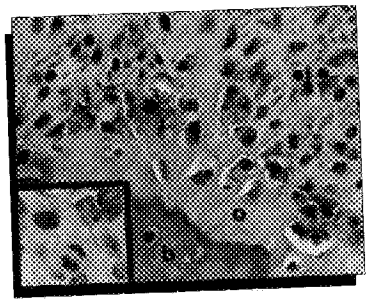

Fig. 6: Microscopically, the osteosarcoma consisted of irregular, malignant cells associated with bone (b) and osteoid (o). Mitotic activity was evident (inset). (H & E, magnification x400, inset: H & E, magnification x400).

dramatically different. This case report stresses the radiologic differences.

Case Report

A 36-year-old, healthy Caucasian female developed a bony, hard, 1.0 cm dome-shaped mass on the buccal aspect of the right maxillary alveolar process in the region of the bicuspids. The lesion, which was asymptomatic and clinically resembled an exostosis, was surgically excised. At age 39, the patient returned with a 2.0 cm, painless, exophytic, bony, hard mass that had occurred in the same site as the previous lesion. It was again excised, and tissue was submitted to the pathology department at a large urban hospital for microscopic examination. A diagnosis of "benign fibro-osseous lesion consistent with fibrous dysplasia" was received. Eight months later, the lesion recurred, growing to 3.0 cm. The patient was referred to an oral surgeon for investigation. On examination, a non-ulcerated, bony, hard, bi-lobed, sessile exophytic mass measuring 3.0 x 1.5 x 1.5 cm was found (**Fig. 2**). The lesion was painless and non-tender to palpation. The teeth were not loose and there was no gingival bleeding. Clinically, the lesion did not exhibit a fusiform growth pattern typical of fibrous dysplasia, nor were the age of onset, the recurrences or the rapid growth rate consistent with fibrous dysplasia. The possibility of malignant disease was considered and the patient was referred for radiographic examination.

Periapical and occlusal radiographs were obtained. The loss of lamina dura and an irregular widening of the periodontal ligament space around the right maxillary cuspid produced a ragged, moth-eaten appearance. Areas of irregular bone destruction and small areas of irregular opacification were noted near the maxillary right cuspid and first bicuspid (**Fig. 3**). The extent and borders of the lesion were not well delineated. Although the teeth were not displaced, a slight resorption of the first bicuspid root was apparent. The antral floor between the roots of the second bicuspid and first molar was not clearly defined (**Fig. 4**), and the maxillary lateral occlusal view demonstrated a subtle change on the buccal aspect. This was better observed under a bright light. A copy film was overexposed to demonstrate irregular bony projections extending perpendicularly from the buccal cortex in the region of the bicuspids (**Fig. 5**). The radiographic appearance was consistent with osteosarcoma or chondrosarcoma. A periapical radiograph obtained seven months earlier demonstrated similar changes to those already described. Tissue sections of the lesion excised eight months previously were also obtained. They showed irregular islands and strands of bone and cartilage, containing and surrounded by polygonal- and spindle-shaped cells that exhibited malignant features. Mitotic figures were present in some areas (**Fig. 6**). A diagnosis of chondroblastic osteosarcoma was reached. The patient underwent a right hemimaxillectomy, followed by a course of chemotherapy (cisplatinum, doxorubicin).

Discussion

Diagnosis is a clinical exercise that always requires appropriate consideration of all available information. This information is derived from the patient history, clinical findings, adjunctive test results and histopathologic findings. Often, it

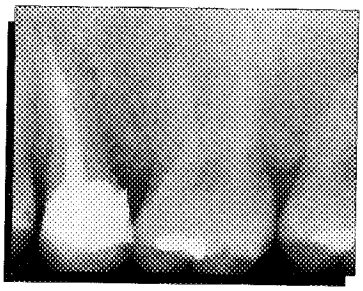

Fig. 7: Typical ground-glass appearance of fibrous dysplasia.

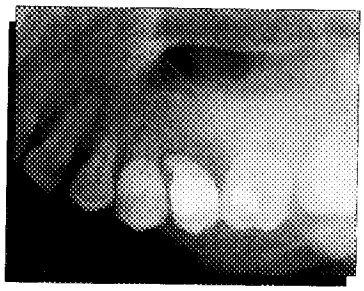

Fig. 8: Typical buccal expansion of maxillary fibrous dysplasia.

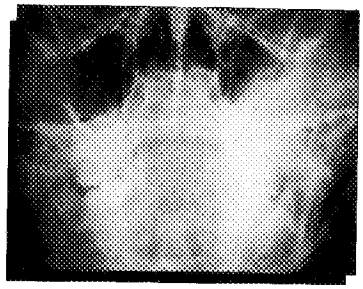

Fig. 9: Typical appearance of maxillary antrum being encroached on by fibrous dysplasia.

is the pathologist who provides the definitive tissue diagnosis. However, accurate microscopic diagnosis is often aided by patient history, clinical findings and test results. In the present case, communicating clinical and radiographic findings to the pathologist may have raised the level of suspicion of more serious disease.

Major differences in the biologic activity of fibrous dysplasia and osteosarcoma are reflected in their radiographic appearances, which vary from patient to patient. In the jaws, fibrous dysplasia typically has a homogeneous, granular, "ground glass" or "orange peel" type of appearance. Some areas of the lesion may have a "finger print" appearance (**Fig. 7**). In contrast, the internal structure of osteosarcoma may have a more variable appearance depending on the amount of bone destruction and production. Typically, irregular bone destruction is observed, producing a "moth-eaten" appearance. This may be superimposed by areas of irregular, variable-sized densities that have lost the normal appearance of trabecular bone (**Figs. 3 and 4**). The change in bone pattern may not be as obvious as in fibrous dysplasia, however, and a critical assessment of general bone pattern is required, as well as a good understanding of normal variation. The borders of fibrous dysplasia are not demarcated by either a radiopaque or radiolucent line. This often causes the lesion to be poorly delineated from the surrounding bone, although it may be quite apparent in some cases. It is usually difficult to identify the borders of osteosarcoma, and there may be areas of abnormal bone extending into areas of normal appearing bone, producing an infiltrating appearance along bone marrow spaces. In the maxilla, fibrous dysplasia typically produces a fusiform, relatively uniform expansion with a smooth, thin cortical surface (**Fig. 8**), while osteosarcoma may rapidly perforate the relatively thin buccal cortex. Once perforation has occurred, a soft tissue mass may be identifiable beyond the normal confines of the bone. Variable amounts of irregular calcification may be seen within this mass. Irregular bony projections, classically described as "sun-ray" spiculation, may be seen perpendicular to the original cortical surface.

Occasionally, this lesion may present as a more solid, irregular mass of calcified material, extending at a right angle to the adjacent cortex (**Fig. 5**). This feature is not invariably observed (Vege et al[8] reported it in only 32 per cent of cases), but its presence should be considered to be ominous. The effect of fibrous dysplasia on the teeth is variable. Often the teeth will appear in their usual positions. However, displacement of teeth or unerupted teeth may be seen and should not be considered unusual. Teeth that are prevented from eruption can give the clinician valuable information regarding the onset of the disorder. Displacement or impaction of teeth is not a feature of osteosarcoma. Resorption is rare,[4] although it is apparent in this case (**Fig. 3**). Usually, the lamina dura or follicle cortex cannot be discerned in fibrous dysplasia. However, the periodontal ligament space is usually apparent unless it is obscured by the thickness of the bone. These latter two structures may reveal changes suggestive of malignant disease. Just as osteosarcoma will rapidly destroy the buccal cortex, it can also destroy lamina dura. Neoplastic tissue may then invade the soft tissue of the periodontal ligament space, producing radiographic widening.

Finally, the effects of these two lesions on surrounding anatomical structures will differ. Fibrous dysplasia in the maxilla will often encroach on the maxillary antrum. This will produce a smaller appearing antral cavity on the affected side. Typically, the lesion will extend into the sinus from the posterior-inferior aspects and extend superiorly and medially to varying degrees (**Fig. 9**). However, the cortical boundaries of the sinus are maintained. Osteosarcoma will cause destruction of antral walls and may fill the antral cavity with a soft tissue mass that may contain calcification. In the mandible, fibrous dysplasia will occasionally cause a superior displacement of the canal, while osteosarcoma will cause destruction of canal boundaries.

Conclusion

This case demonstrates the need for an integrated approach to accurate diagnosis. Because dentists routinely expose and interpret their patients' radiographs, they must be aware of the radiographic features of aggressive disease. The ability to recognize normal, a variation from normal, and abnormality requires experience and a thorough evaluation of each radiograph exposed. General dentists must also realize that microscopic diagnosis is subject to error and often requires historical, clinical, and radiographic correlation to be accurate. ■

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